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Posterior Lung Herniation in Pulmonary Agenesis and Aplasia: Chest Radiograph and Cross-Sectional Imaging Correlation

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Objective: To describe the anatomic locations and imaging features of posterior lung herniation in unilateral pulmonary agenesis and aplasia, focusing on radiograph-CT/MRI correlation.

Materials and Methods: A total of 10 patients (seven with pulmonary agenesis and three with pulmonary aplasia, male: female = 1:9, mean age 7.3 years, age range from 1 month to 20 years) were included. Chest radiographs (n = 9), CT (n = 9), and MRI (n = 1) were reviewed to assess the type of lung underdevelopment, presence of anterior and posterior lung herniation, bronchus origin, supplying artery, and draining vein of the herniated lung.

Results: Pulmonary agenesis/aplasia more commonly affected the left lung (n = 7) than the right lung (n = 3). Anterior lung herniation was observed in nine of the 10 patients. Posterior lung herniation was observed in seven patients with left pulmonary agenesis/aplasia. Two patients showed posterior lung herniation crossing the midline but not beyond the aorta, and five patients showed the posteriorly herniated right lower lobe crossing the midline to extend into the left hemithorax farther beyond the descending thoracic aorta through the space between the esophagus and the aorta. This anatomical configuration resulted in a characteristic radiographic finding of a radiolucent area with a convex lateral border and a vertical medial border in the left lower lung zone, revealing a tongue-like projection on CT and MRI.

Conclusion: Posterior lung herniation occurs in unilateral left lung agenesis/aplasia. Approximately 70% of the cases of posterior lung herniation reveal a unique radiolucent tongue-like projection in the left lower lung zone on imaging studies, which is caused by the extension of the posteriorly herniated right lung farther beyond the descending aorta.

Keywords: *Pulmonary agenesis; Pulmonary aplasia; Posterior lung herniation; Chest radiography; CT*

INTRODUCTION

Pulmonary underdevelopment is a rare developmental anomaly that is categorized as follows: 1) pulmonary agenesis: the complete absence of the lung parenchyma, bronchus, and pulmonary vasculature, 2) pulmonary aplasia: the absence of the lung parenchyma and pulmonary vasculature, while the rudimentary blind-ending bronchus is present, 3) pulmonary hypoplasia: the presence of a bronchus and rudimentary lung parenchyma with a reduction in the number and size of the alveoli, airways, and pulmonary vasculature [1-3]. Pulmonary agenesis

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and aplasia (agenesis/aplasia) may occur bilaterally or unilaterally. However, since bilateral pulmonary agenesis/aplasia is fatal, nearly all reported cases of living patients have unilateral pulmonary agenesis/aplasia [1,2].

The radiographic finding of unilateral pulmonary agenesis/aplasia is a small, completely opaque, ipsilateral hemithorax, which is associated with mediastinal shifting to the ipsilateral side, and ipsilateral rib crowding [2,3]. In most cases of unilateral pulmonary agenesis/aplasia, the mediastinum rotates and shifts toward the ipsilateral direction, which causes the contralateral lung to over-expand, resulting in lung herniation. This finding can lead to the misinterpretation that the lung parenchyma arising from the ipsilateral thoracic cavity exists. This may be mistaken for partial atelectasis or pneumonic consolidation of the ipsilateral lung, thereby making an accurate diagnosis challenging. CT and MRI can be valuable in the accurate diagnosis of accompanying lung herniation.

Anterior herniation of the contralateral over-expanded lung in unilateral pulmonary agenesis/aplasia is well-described in the literature [1-6]. However, there are only a few case reports of posterior herniation [3-6]. Even in these reports, details on the exact anatomical configuration of the contralateral lung crossing the midline to the ipsilateral hemithorax have not been well-described.

Therefore, the purpose of this study was to describe the anatomic locations and imaging features of posterior lung herniation in unilateral pulmonary agenesis/aplasia, focusing on the radiograph-CT/MRI correlation.

MATERIALS AND METHODS

This retrospective study was approved by the Institutional Review Board of Seoul National University Hospital (J-2010-118-1166) and Seoul National University Bundang Hospital Institutional Review Board (B-2006-621-102). Informed consent was waived because of the retrospective nature of the study. All confirmed cases of unilateral pulmonary agenesis/aplasia between January 1991 and December 2019 were included in the study. We reviewed the medical records of each patient and recorded their age, sex, clinical presentation, and combined congenital anomalies. All available chest radiography (n = 9), CT (n = 9), and MRI (n = 1) were reviewed by two pediatric radiologists (with 31 and 10 years of experience, respectively). This study retrospectively reviewed images obtained over 28 years and acquired using various protocols. Chest radiography consisted of posteroanterior or anteroposterior projections with the patient in a supine or standing position, depending on the age and clinical condition of the patients.

The presence of a rudimentary bronchus was assessed using cross-sectional imaging studies (CT and MR) to evaluate the type of lung underdevelopment (agenesis vs. aplasia). Other imaging findings analyzed in this study were as follows: 1) site of the absent lung parenchyma (right vs. left), 2) presence of an anterior lung herniation (lung crossing the midline and protruding to the contralateral side in the anterior thoracic cavity), and 3) presence of a posterior lung herniation (lung crossing the midline and protruding to the contralateral side in the posterior thoracic cavity). The anatomical positions of the herniating lungs are also described. For cases with posterior herniation, we evaluated whether the pulmonary parenchyma extended beyond the aorta (Fig. 1). The origin of the bronchus, supplying artery, and draining vein of the herniated lung were also determined.

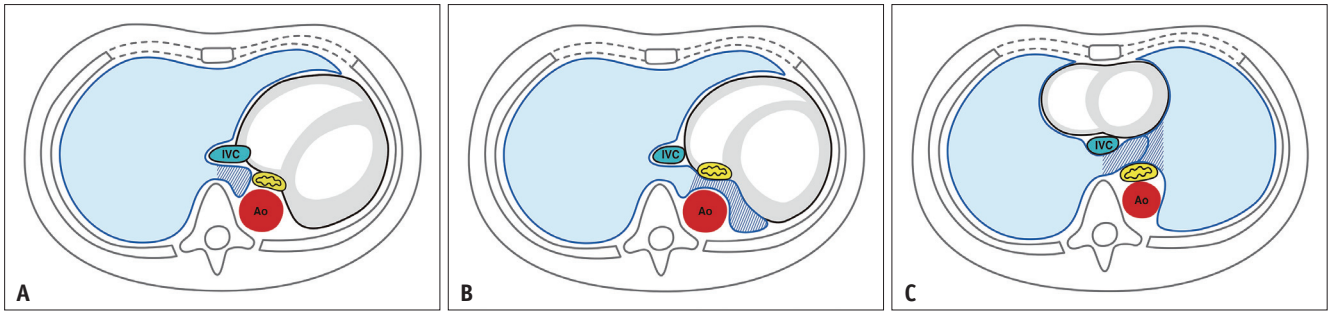

Fig. 1. Schematic drawings of posterior lung herniation in left pulmonary agenesis (A, B) and horseshoe lung (C).
A. Posterior right lung herniation not beyond the Ao (area marked in darker blue). **B.** Posterior right lung herniation beyond the Ao (areas marked in darker blue) passes between the esophagus (yellow structure) and the Ao. **C.** Horseshoe lung with a posteriorly herniated lung (areas marked in darker blue) cross the posterior mediastinum anteriorly to the esophagus (yellow structure). Ao = aorta, IVC = inferior vena cava

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RESULTS

Clinical Findings

Ten patients were included in the retrospective analysis. Nine patients were women. At the time of initial diagnosis, their ages ranged from 1 month to 20 years (mean age 7.3 years). Table 1 summarizes the clinical features of these patients.

Imaging Findings

Table 2 shows the incidence of pulmonary herniation according to location. Posterior lung herniation was identified in all seven patients with left pulmonary agenesis/aplasia. None of the patients with right pulmonary agenesis showed a posterior lung herniation. In five of the seven patients with a posterior lung herniation, the posteriorly herniated right lower lobe not only crossed the midline but also extended to the left thorax farther beyond the aorta ("posterior lung herniation beyond the aorta") (Fig. 2). In the remaining two patients with a posterior lung herniation, the right lung crossed the midline to the left thorax but not beyond the aorta ("posterior lung herniation not beyond the aorta") (Fig. 3). In six of the seven patients with posterior lung herniation, posterior lung herniation not beyond the aorta appeared only below the carina (Fig. 3). In the remaining patient, posterior lung herniation not beyond the aorta occurred both above and below the carina (Fig. 2). Above the carina, the posteriorly herniated right lung crossed the midline through the space between the trachea and the vertebral body at the midline. Below the carina, the posteriorly herniated right lungs crossed the midline through the space between the inferior vena cava or the right inferior pulmonary vein and the vertebral body. This resulted in the deepening of the azygoesophageal recess. A posterior lung herniation beyond the aorta was visible below the carina in all five patients. All the posteriorly herniated right lungs that extended beyond the aorta passed through the space between the esophagus and the aorta, revealing a tongue-like projection on CT or MRI (Fig. 2). On chest radiography, the radiolucent areas with a convex lateral border and a vertical medial border in the left lower lung field below the pulmonary hilum were observed in all patients with posterior lung herniation beyond the aorta.

Anterior lung herniation was observed in all the patients, except in one patient with left pulmonary agenesis. Based on the CT and MRI findings, the anatomical locations of the anterior lung herniation were similar in the nine patients. Anterior lung herniation occurred throughout the length of the thoracic cavity, from the thoracic inlet or thymus to the diaphragm. The anteriorly herniated lung crossed the midline through the space between the sternum and the heart (Fig. 4). The thymus was pushed toward the contralateral thoracic cavity by an anteriorly herniated lung.

Table 1. Clinical Features of the Patients

Case No.	Age	Sex	Clinical Presentation	Presence of Combined Anomaly	Imaging Study	Agenesis vs. Aplasia
1	13 y	Female	Incidental†	No	CR, MR	Left pulmonary aplasia
2	14 y	Female	Incidental†	No	CR, CT	Left pulmonary aplasia
3	10 m	Female	Failure to thrive	No	CR, CT	Left pulmonary aplasia
4	10 y	Female	NA	Renal agenesis (left), hemivertebra, rib fusion	CR, CT	Left pulmonary agenesis
5	20 y	Female	Incidental†	No	CT	Left pulmonary agenesis
6	13 y	Female	Incidental†	No	CR, CT	Left pulmonary agenesis
7	5 m	Female	Dyspnea	Spine & rib abnormality*, CDH	CR, CT	Left pulmonary agenesis
8	1 m	Male	Respiratory difficulty	Tracheal stenosis	CR, CT	Right pulmonary agenesis
9	6 m	Female	Respiratory difficulty	Tracheal stenosis	CR, CT	Right pulmonary agenesis
10	1 y	Female	NA	No	CR, CT	Right pulmonary agenesis

*Butterfly vertebra, spine fusion, partial agenesis of the spine, and rib fusion, †Four patients without specific respiratory symptoms were diagnosed coincidentally during a general health examination owing to abnormal chest radiography findings or abnormal breathing sounds. CDH = congenital diaphragmatic hernia, CR = chest radiography, m = months, NA = not available, y = years

Table 2. Incidence of the Lung Herniation by Location

	Anterior Lung Herniation (%)	Posterior Lung Herniation	
		Beyond the Aorta (%)	Not Beyond the Aorta Only (%)
Right pulmonary agenesis (n = 3)	3 of 3 (100)	0	0
Left pulmonary agenesis/aplasia (n = 7)	6 of 7 (85.7)	5 of 7 (71.4)	2 of 7 (28.6)

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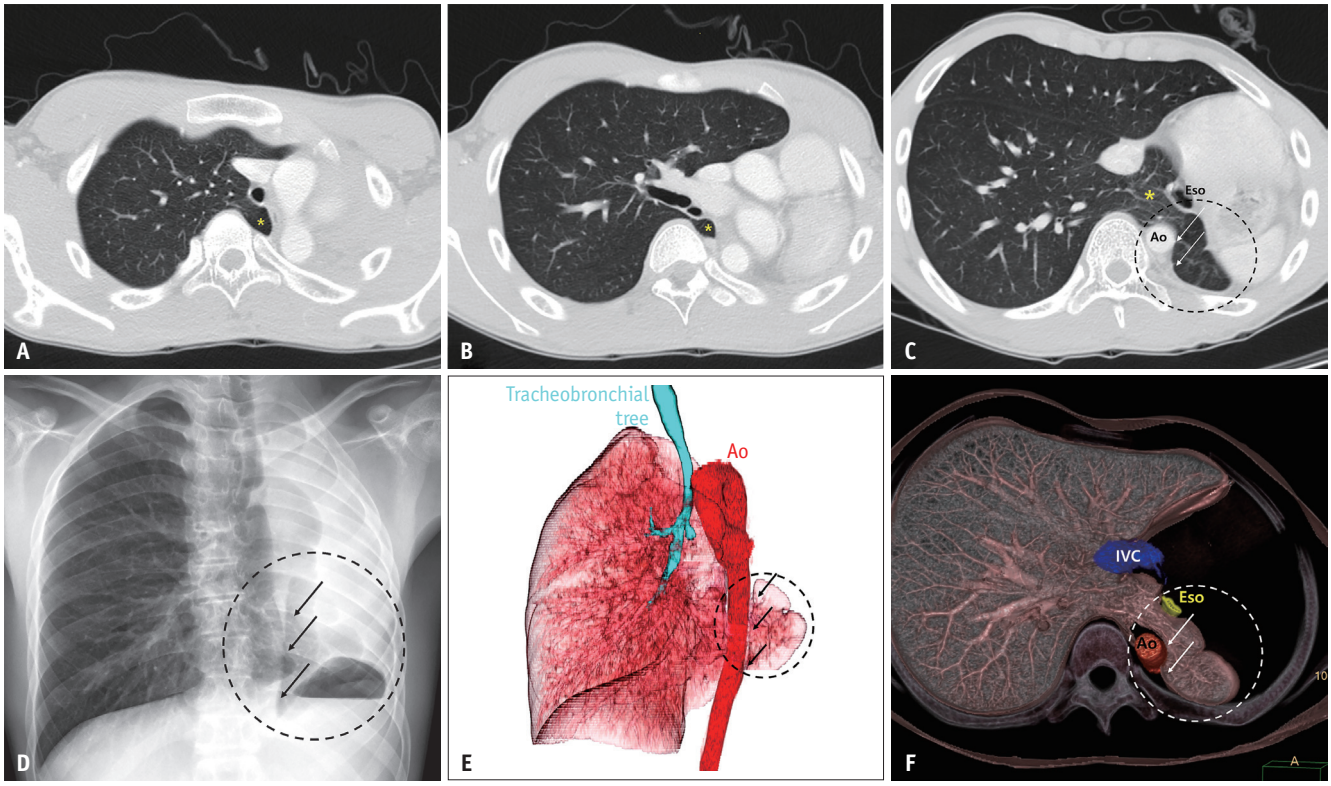

Fig. 2. Posterior lung herniation extending beyond the Ao associated with anterior lung herniation in a 14-year-old girl with left lung aplasia.

A-C. As with anterior lung herniation, posterior lung herniation not beyond the Ao (*) is demonstrated throughout the whole thoracic cavity. **C.** The posteriorly herniated right lung passes between the Eso and the Ao and extends beyond the Ao, resulting in tongue-like projection (circle with dotted line). **D-F.** The posterior lung herniation beyond the Ao (circle with dotted line) is shown with a convex lateral border and a vertical medial border in the left lower lung field on chest radiography and 3D volume rendering images. The discrete medial vertical line (arrows) is formed by the Ao. **B, E.** The rudimentary left main bronchus is demonstrated in the CT axial view and 3D volume rendering images. Along with mediastinal shifting, mechanical overstretch occurs in the tracheobronchial tree, and the AP dimension of the trachea is reduced. Ao = aorta, AP = anteroposterior, Eso = esophagus, IVC = inferior vena cava, 3D = three-dimensional

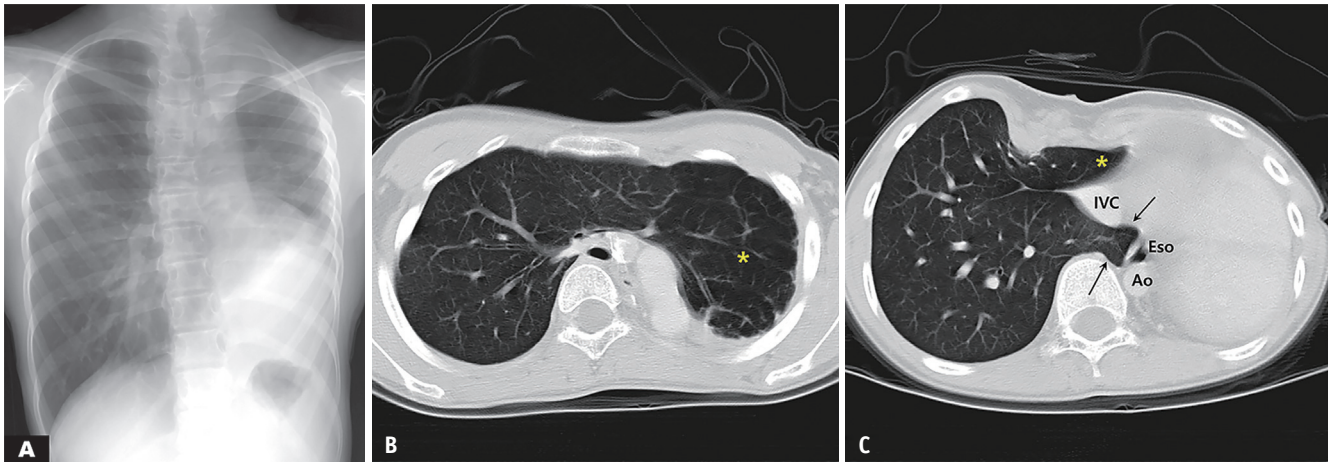

Fig. 3. Posterior lung herniation not extending beyond the Ao associated with anterior lung herniation in a 13-year-old girl with left pulmonary agenesis.

A. Chest radiography shows the left elevated diaphragm and rib crowding. Extensive anterior lung herniation causes a radiolucency in the left thorax. **B, C.** Anterior lung herniation (*) is observed in the upper **(B)** and lower **(C)** thoracic cavities, whereas posterior lung herniation (arrows) is observed in the lower thoracic cavity below the carina. The posteriorly herniated lung crosses the midline between the IVC and the vertebral body and deepens the azygoesophageal recess. The posteriorly herniated right lung does not extend to the left side of the Ao. Along with mediastinal shifting, mechanical overstretch occurs in the tracheobronchial tree, and the AP dimension of the trachea is reduced. Ao = aorta, AP = anteroposterior, Eso = esophagus, IVC = inferior vena cava

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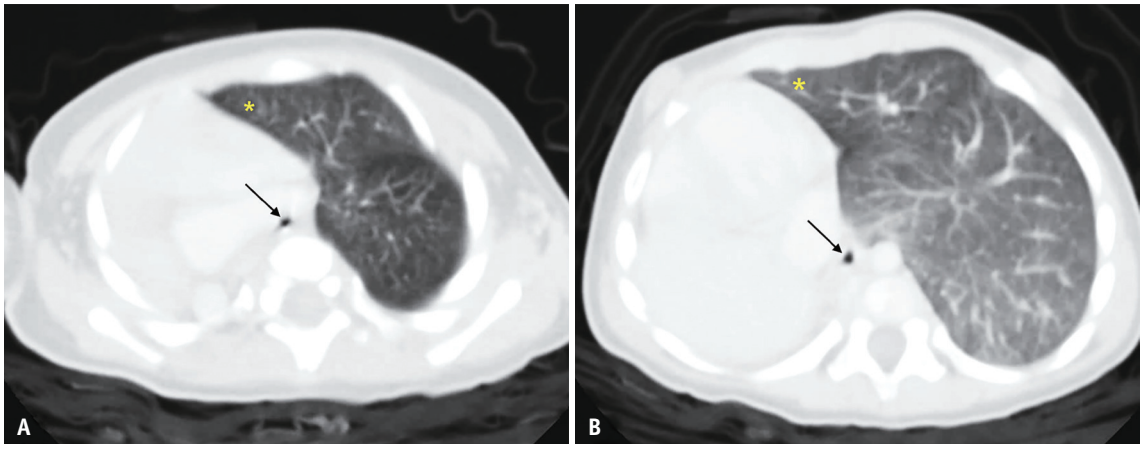

Fig. 4. Anterior lung herniation in a 6-month-old girl with right pulmonary agenesis.
A, B. The right lung parenchyma is absent, and the mediastinum shifting to the right hemithorax is shown. The anteriorly herniated lung (*) crosses the midline through the space between the sternum and the heart and is consistently observed in the upper **(A)** and lower **(B)** thoracic cavity. The small diameter of the trachea (arrows) suggests an accompanying congenital tracheal stenosis.

Table 3. Detailed Description of Anatomic Structures Constituting the Herniated Lung

	Bronchus Origin	Supplying Artery	Draining Vein
Anterior lung herniation	RUL & RML in left pulmonary agenesis/aplasia LUL in right pulmonary agenesis	RPA in left pulmonary agenesis/aplasia LPA in right pulmonary agenesis	RSPV in left pulmonary agenesis/aplasia LSPV in right pulmonary agenesis
Posterior lung herniation not beyond the aorta	RUL (above the carina) RLL (below the carina)	RPA	RSPV (above the carina) RIPV (below the carina)
Posterior lung herniation beyond the aorta	RLL	RPA	RIPV

LPA = left pulmonary artery, LSPV = left superior pulmonary vein, LUL = left upper lobar bronchus, RIPV = right inferior pulmonary vein, RLL = right lower lobar bronchus, RML = right middle lobar bronchus, RPA = right pulmonary artery, RSPV = right superior pulmonary vein, RUL = right upper lobar bronchus

Table 3 summarizes the detailed anatomic structures of the herniated lung. The anatomic structures could not be assessed in one patient using MRI, and this necessitated contrast enhancement for the image analysis. There was no aberrant systemic arterial supply or anomalous pulmonary venous return in the herniated lungs.

DISCUSSION

The herniation of the contralateral lung into the ipsilateral thoracic cavity has been well-established in patients with unilateral pulmonary agenesis/aplasia [1-6]. However, most cases of lung herniation reported in unilateral pulmonary agenesis studies were anterior, while posterior herniation has not been well-described. We separately described the two types of posterior lung herniation: "herniation not beyond the aorta" and "herniation beyond the aorta." To the best of our knowledge, only four reports have described posterior lung herniation in unilateral pulmonary agenesis [3-6]. Three of these reports showed posterior lung herniation beyond the aorta. Interestingly, all three cases had left pulmonary agenesis, which is consistent with our results. The descending thoracic aorta is usually located in the left paramedian area of the posterior mediastinum. This localization may be consistent in patients with pulmonary agenesis/aplasia. The descending thoracic aorta would have to be dislocated to the right side in a patient with right pulmonary agenesis for posterior lung herniation to develop. Therefore, we hypothesized that lateralization may be a factor that prevents posterior lung herniation in right pulmonary agenesis/aplasia.

When referring to the CT images in the published reports, the anatomic location where the posterior lung herniation extended beyond the aorta was the space between the esophagus and the descending thoracic aorta in all three cases, which was consistent with our results [3,4,6]. According to our findings, the location where the posterior lung herniation beyond the aorta occurred in

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unilateral pulmonary agenesis was anatomically different from where the lung crossed the posterior mediastinum in cases of a horseshoe lung (Fig. 1). In the horseshoe lung, the posteriorly herniated lungs cross the posterior mediastinum in front of the esophagus, between the heart and esophagus, and below the carina [7-12]. Considering that a horseshoe lung can result from non-separation of the lung bud arising from the ventral aspect of the tubular foregut, both lungs may fuse on the ventral side of the esophagus [13]. With information on the anatomical locations where the lungs cross the posterior mediastinum in the horseshoe lung and left pulmonary agenesis with posterior lung herniation, radiologic differentiation of the two abnormalities can be easier.

Posterior lung herniation can also occur in patients undergoing pneumonectomy. However, none of these patients in the previous reports showed a posterior lung herniation crossing the descending thoracic aorta [14-17]. These findings allow us to assume that "posterior lung herniation beyond the aorta" may not occur when the left lung absence is not congenital (which is, after a left pneumonectomy). Additionally, this anatomical finding of posterior lung herniation across the aorta may be a characteristic finding of a congenitally absent left lung, such as that observed in left pulmonary agenesis or aplasia.

A posteriorly herniated lung that crosses the aorta to the contralateral side can result in a unique radiographic finding: the radiolucent area of a tongue-like projection with a convex border and a vertical medial border on the left lower lung field below the pulmonary hilum. This finding was also reported previously [4]. On comparing chest CT/MRI and radiographic findings, we observed that the aorta forms the vertical medial border of the radiolucent area, and the herniated lung through the space between the aorta and esophagus showed a tongue-like projection on CT or MRI. These imaging findings were identified for all the cases in our study and previously published reports of posterior lung herniation beyond the aorta [3,4,6,18]. In unilateral pulmonary agenesis/aplasia, the affected hemithorax may appear radiolucent on chest radiography due to herniation of the contralateral lung. As a result, it can be misinterpreted as pneumonic consolidation or partial atelectasis of the affected hemithorax, which may delay an accurate diagnosis. However, posterior lung herniation beyond the aorta showed characteristic imaging findings on chest radiography. For unilateral pulmonary agenesis with posterior lung herniation beyond the aorta, knowledge of the aforementioned radiographic findings may help direct the appropriate diagnosis even with only plain radiography.

Our study had some limitations. First, a few cases were included in our analysis owing to the rare occurrence of pulmonary agenesis/aplasia. To overcome this limitation, a literature review was performed to confirm the comparability and consistency of our findings with those of previous reports. Second, this was a retrospective study; therefore, clinical information was obtained only from medical records. This may have caused inadequate clinical information regarding the clinical symptoms or associated anomalies, which may not have been available in some cases. In addition, since this study retrospectively reviewed images performed in a few cases over 28 years, a comprehensive analysis is challenging due to different imaging acquisition methods and patients of various ages.

In conclusion, posterior lung herniation in unilateral lung agenesis/aplasia occurred only in patients with left pulmonary agenesis/aplasia, while anterior lung herniation occurred in both the right and left pulmonary agenesis/aplasia. Two types of posterior lung herniation were observed: a herniation crossing the midline, but not beyond the aorta, and another herniation crossing the midline farther beyond the aorta. When the right lower lobe herniates beyond the descending thoracic aorta to the left thorax, the herniated lobe passes through the space between the esophagus and the aorta. This results in the characteristic imaging findings of a radiolucent area with a convex lateral border and a vertical medial border in the left lower lung field on plain radiography and a tongue-like projection on CT and MRI.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Author Contributions

Conceptualization: Ji Young Kim, Woo Sun Kim. Data curation: all authors. Formal analysis: Ji Young Kim, Woo Sun Kim, Kyung Soo Lee. Funding acquisition: Ji Young Kim. Investigation: all authors. Methodology: all authors. Project administration: Ji Young Kim, Woo Sun Kim, Kyung Soo Lee. Resources: all authors. Software: Ji Young Kim, Woo Sun Kim. Supervision: Ji Young Kim, Woo Sun Kim. Writing—original draft: Ji Young Kim, Woo Sun Kim, Kyung Soo Lee. Writing—review & editing: all authors.

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